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CASE REPORT

Case Report and Literature Review: Intraoperative Injury to Cisterna Chyli During an Elective Pancreatic Resection Leads to Chylous Ascites and a Poor Outcome

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Background: Chylous Ascites is a rare condition. It develops as a result of iatrogenic injury to the cisterna chyli, thoracic duct, or their major tributaries. It leads to an increase in chyle release. Subsequently, obstruction of lymphatic return from abdomen. It is more prevalent after major abdominal procedures involving retro-peritoneum.

Case Presentation: A 65 years old female patient was admitted with a history of pain at epigastric and left upper abdominal quadrant via the emergency department at King Hussein Medical Centre. She was planned for elective Whipple's procedure for cancer head of pancreas. This case report considers the impact of intraoperative injury to major lymphatic system leading to chylous ascites and death following her pancreatic resection. Retrospective evaluation of her records revealed an intraoperative iatrogenic injury to the cisterna chyli had occurred during Whipple's Procedure. The injured lymphatic duct was discovered and repaired intraoperatively. The chyle drainage was resolved temporarily after 3 weeks of conservative management. Despite its resolution, it recurred after massive upper GI bleeding, which was managed operatively after failed endoscopic and embolization. The patient failed to recover and died as a direct result of her poor general condition due to the effects of an iatrogenic injury of Cisterna Chyli. We report this case of intraoperative injury to the cisterna chyli, subsequent development of chylous ascites, and its post-operative sequelae. This article considers, even if discovered and repaired intraoperatively, the impact of such injury on development of postoperative chylous ascites, nutritional insufficiency, poor prognosis, and subsequent death.

Conclusion: Chylous ascites is a rare complication of Whipple's Procedure. It usually progresses due to iatrogenic operational trauma to a significant lymphatic channel. Even if such injury to cisterna chyli is discovered and managed intraoperatively, it carries a poor prognosis and subsequent death.

Keywords: Whipple's Procedure, WP, Chylous ascites, CA, Pancreatico-Duodenectomy, PD, Cisterna chyli, CC

Background

Chylous Ascites (CA) following Pancreatico-Duodenectomy (PD) is an uncommon event.[1] Other studies have largely considered post-operatively diagnosed CA.[1] This paper explores post-operative sequelae of the damage to the Cisterna Chyli (CC) noted intraoperatively and the development of CA despite an immediate treatment being commenced.

Case Presentation

We report a case of intraoperative traumatic injury to CC during Whipple's Procedure (WP) in a 65 years old female patient at King Hussein Medical Center (KHMC). The patient was known to have diabetes mellitus, hypertension, and COPD. She was admitted to the KHMC Accident and Emergency department on the 24th of September, 2005. She had a one-week history of epigastrics and pain at the left upper abdominal quadrant. On presentation, our patient was cyanosed and pale but not jaundiced. The patient had abdominal tenderness and guarding in the right upper quadrant on

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deep palpation but no mass could be felt. The remaining physical examination was unremarkable. Investigations revealed a hematocrit of 36%, a white cell count of 14,000/mL. Her pulmonary function test was poor. Renal function, serum electrolyte, serum glucose, serum amylase, liver function, routine coagulation studies, and urine analysis were all normal. Abdominal ultrasound and CT scans were performed and showed an enlarged head of pancreas with minimal peri-pancreatic fluid and distended gallbladder, resulting in a diagnosis of acute pancreatitis. A second ultrasound and EUS done on the same day confirmed the presence of a 2cm hypo-echoic lesion in the head of pancreas and a follow-up FNA confirmed the presence of a ductal adenocarcinoma. The dangers of the process were discussed with the patient and her family, and she experienced a Whipple's Procedure on the 18th October. Intraoperatively, an injury to the cisterna chyli occurred. That injury was discovered intraoperatively, and sewn with vicryl stitches and the patient was transferred to the ICU. The patient stayed three days before being transferred to the ward. By the fifth day postoperatively her abdominal drain was producing between 1000cc-1200cc per day. This output was cloudy, milky, and turbid in appearance. A clinical diagnosis of CA was made due to an injury to cisterna chyli. Standard blood tests including a complete blood count, electrolytes, LFTs, total protein, albumin, LDH, TG, cholesterol, amylase, and lipase were requested, and fluid from the drain was also sent for cell count, culture, Gram stain, triglycerides, total protein concentration, albumin, glucose, lactate dehydrogenase, amylase, and fluid cytology were performed. Triglyceride values were 350 mg/dL and total protein 5g/dL were in keeping with the clinical diagnosis of CA. The patient was managed conservatively with TPN, medium-chain triglycerides (MCT) diet and somatostatin for 3 weeks before the chyle ceased draining. Although chyle draining stopped, she was unwell and fragile. Then, the patient developed massive upper GI bleeding on the 9th of November. She was resuscitated and transferred to ICU where she was further investigated and infused with packed RBCs and FFP. However, she underwent an upper endoscopy, an angiogram, and an embolization, but they failed to stop her bleeding. An emergency laparotomy was performed to control the bleeding. However, it was successful, the chyle draining returned and her condition deteriorated and died later on the 7th of December, with a formal immediate cause of death given by ICU doctor, as type II acute respiratory failure and her CA post Pancreatico-Duodenectomy as the primary leading cause.

Discussion

Out of the 30 patients who had undergone pancreatic resections at KHMC between 2002 and 2006, only one case (3.3%) developed CA post Pancreatico-Duodenectomy (PD). The majority of our 30 patients required pancreatic resection due to malignancy (80%), of which the majority were diagnosed with pancreatic head adenocarcinoma. Overall, 70% of patients had a PD and 10% underwent a Distal Pancreatectomy (DP). However, 20% of cases had DP for benign causes. Chylous Ascites is a rare condition.[1] Frequently, it develops as a result of iatrogenic injury to the cisterna chyli, thoracic duct, or their major tributaries. Chylous ascites lead to increase chyle release. Subsequently, obstruction of lymphatic return from the abdomen. Chyle production is drained via lymphatics which drain the viscera of the abdomen and retro-peritoneum and combine the ascending lymphatic trunks from the lower extremities and the pelvis. These ascending trunks form a saccular dilatation called the "cisterna chyli" (CC), which lies in front of the first and second lumbar vertebrae. Also, it drains lymph into the thoracic duct inside the thoracic cavity. Chylous Ascites develop when there is a disruption of this system, which can occur as a result of traumatic injury or obstruction from benign or malignant causes which can result in chylo-thorax as well as CA.[1] Chylous Ascites is more prevalent after major abdominal procedures involving retro peritoneum.[2] Many classifications of CA have been published based on the physical appearance and biochemical/bacteriological constituents.[1,2] Typically, triglyceride levels of more than 200 mg/dl in the ascitic fluid are helpful in confirming the diagnosis. Others as specific gravity greater than 0.012, a fat content of 4–40 g/L, alkaline pH, and the presence of sterile chyle.[1,2] Extensive retroperitoneal dissection in PD may injure lymphatics, leading to leaks of chyle which might produce a CA leading to the patent proximal thoracic duct and extensive collateral lymphatics. CA is a milky appearing peritoneal fluid that is rich in TG caused by the leaking of thoracic or intestinal lymph into the abdominal space.[3] CA following abdominal procedure is uncommon and its treatment is deficiently managed, although there have been some recent studies showed an incidence may reach 11%.[3] An Iatrogenic injury and disruption to intra-abdominal CC lymphatics tributaries leaks are most likely cause.[4] Clinically, CA presents as painless progressive abdominal distention, occurring over weeks to months depending upon its cause.[3] On the other side, Chyle consists of intestinal lymphatic fluid enriched after feeding with

fat-soluble vitamins and LCT incorporated in chylomicrons into blood stream. Presence of fat explains the cloudy appearance of chyle. Because lymphatic fluid contains lymphocytes and immunoglobulin, loss of the fluid results in lymphocytopenia, which renders patients susceptible to infection-related complications.[1] However, acute presentation may occur in patients who had abdominal or thoracic surgery. Patients may increase their weight, become dyspnoeic, as a result of increase in abdominal pressure. Other features may include diffuse abdominal pain, fever, nausea, early satiety diarrhea, reduced absorption of fat by the intestine, malnutrition, edema and night sweats.[1] Nevertheless, due to wide range and lack of specificity in the symptoms, the majority of diagnoses are not made before the appearance of chyle via an operative drain or following diagnostic paracentesis. Fundamental complaints such as anorexia, weakness, and general feeling of discomfort, are also very common but imprecise.[1] Usually, the chyle has a overcast and confused appearance, in difference to the yellow and transparent appearance of ascites as the outcome of cirrhosis or portal hypertension. The triglyceride levels in the ascetic fluid are critical in describing CA and are usually significant if found to be above 200 mg/dL, despite that some authors utilize a cut-off value of 110 mg/dL.[3] The total protein content varies resting upon the underlying cause, ranging between 2.5 to 7.0 g/dL. The ratio of Serum to Ascetic collection Albumin Gradient (SAAG) should be measured by subtracting the ascetic fluid value of albumin from the serum albumin value to decide if that ascetic fluid is due to portal hypertension or other pathology.[4] Ascetic fluid sent for TG levels, the cell count, culture and sensitivity, total protein, albumin, blood sugar, LDH, amylase, for gram stain and cytology. Standard blood tests as a CBC, electrolytes, LFT, total protein, albumin, LDH, TG, cholesterol, amylase, and lipase should be requested. Other tests might be performed should be founded on medical setting. Lymphangiography is the gold average in significant patients of obstruction, however it carries numerous likely complications, such as fat embolism, tissue necrosis, and hypersensitivity. Nevertheless, these responses should be achieved as soon as possible.[5] In the setting of iatrogenic causes CA, abdominal scans help in determining the extent and localization of leaked fluid, particularly if there is a suspicion of thoracic duct injury. Lymphangiography and lymphoscintigraphy, can assist in detecting abnormal retroperitoneal nodes, leaks from dilated lymphatics, fistulation, and patency of the thoracic duct.[5] Only a few studies have addressed specific treatments aimed at decreasing the development of ascites. Based upon the limited published data, a reasonable initial approach for patients in whom the cause cannot be found or those who do not respond to initial therapy is to recommend a high protein and low fat-diet with medium-chain triglycerides (MCT) and limited use of of long-chain triglycerides (LCT).[6] Restrictions of LCT is to avoid their metabolic products, mono-glycerides and free fatty acids (FFA), which are transported into the intestinal lymphatics to the chylous blood via thoracic duct as chylomicrons. While the MCTs are absorbed directly into intestinal cells and transported as FFA and glycerol directly to the liver via the portal vein. So, a low fat-diet contains LCT and high MCT supplementation decreases the production and flow of chyle.[6] MCT oil is commercially available in Jordan. The usual initial oral adult dose of MCT oil as a nutritional supplement is one tablespoon every 6 to 8 hours daily, which was given for our patient. Usual side effects were include nausea, sometimes vomiting, abdominal discomfort, and loose frequent defecation. Several other authors have been described in their case reports using Orlistat, which is a reversible inhibitor of gastric and pancreatic lipases.[7] Also it helps cirrhotic patients having CA to minimize ascites and TG values. Somatostatin and octreotide have been used successfully to treat chylous effusions in patients with yellow nail syndrome and lymphatic leaks due to abdominal and thoracic surgery.[7] Parenteral nutrition is used in patients who do not respond to the above methods. Since enteral feeding stimulates intestinal lymph flow. Some authors recommend both somatostatin and hypodermic octreotide for dealing with Chylous Ascites.[8] In the past, perito-venous shunts were associated with a high rate of withdrawals, including infection, disseminated intravascular coagulation (DIC), electrolyte imbalance, small intestinal obstruction and the occurrence of air embolism. Also, high viscosity of the chyle results in a significant rate of shunt occlusion, thus they are no longer use.[8] On the other hand, Van der Gaag considered TG content as highly relevant believing that the TG level in lymph is possibly related to the nutritional status of the patient.[9] Malnutrition is not uncommon in patients requiring a resection of the pancreas, which might result in decreased levels of TG in serum and lymph. This, therefore, will result in under diagnosis of Chylous ascites if clinicians depend on cut-off of 1.2 mmol/L for TG level in the drain output. In addition, to establish a diagnosis of CA, the TG content of drain output needs to exceed the serum TG level.[8] It might be suggested whether or not a drain output below 100 mL/day, despite a TG level greater than 1.2 mmol/L, should be regarded as CA or merely a chyliform type of effusion. Van der Gaag also demonstrated that female patient and chronic pancreatitis to be independently associated with development of isolated CA, and he mentioned that the incidence was around 11% and

with clinically significant CA.[9] Reoperations to repair injured lymphatic duct were not performed nor insertion of a peritoneo-venous shunt in our case. We gave our patient an LCT-restricted diet rich in MCT after an established diagnosis of CA with TPN. Malik recently suggested using TPN as a first-line measurement for CA after PD.[10] While other authors recommend using TPN as a second-line for treatment of refractory cases of isolated CA. Those authors related that TPN is expensive and requires insertion of central venous line, which harbors a risk of line sepsis and septicemia. However, enteral feeding maintains intestinal mucosa integrity, in contrast to TPN. Most reports of chyle leak as a postoperative complication have included patients who underwent retroperitoneal dissection of lymph node, distal spleno-renal shunts, abdominal aortic aneurysm repair, or hepatic transplant surgery.[11] A small number of case reports have reported patients who had pancreatic resection. Information for chyle leaks following resection of pancreas are largely unavailable and the majority based on case reports or observational studies mentioned in the literature.[10] Assumpcao studied 3532 patients who underwent pancreatic resections, 47 (1.3%) developed a chyle leak following surgery.[12] Complications associated with the chyle leak were mentioned as abscess in 4.3%, pancreatic fistula 4.3%, malnutrition (serum albumin <3.5 mg/dl) was 91.5%, peritonitis developed in 6.4% of patients, and sepsis was in 12.8%.[12] Assumpcao also reported that the median time to disappearance of the chyle drainage was 13 days (range 8 to 27 days). This was not significantly different in comparison to those managed conservatively with TPN, which resolved 15 days (range, 9 to 28 days). Meanwhile, those who required more aggressive management (eg lymph scans or re-operation) required a median of 2 months for the chyle leak to resolve. He concluded that chyle leaks were a rare and severe consequence, with a bad prognosis following pancreatic resection with 1.3% incidence rate. In general, chyle leaks are successfully managed with TPN with no adverse impact on outcome. Patients with CA, however, had a more prolonged duration of their recovery and a worse long-term survival. However, Malik et al reported a high incidence of chyle leaks postoperatively in pancreatic-duodenal malignancy.[10] That high rate was related into early enteral nutrition among those patients. He reported that a total of 6.7% of patients were identified as having a significant chyle leak. The maximum volume of chyle drained daily ranged from 1.4 to 3 L.[10] Walker reported the same complication and considered that CA following PD might be due to the obstruction or malignant infiltration of lymphatic ducts with then the development of lymphatic fistula, which communicates with the peritoneal space. Iatrogenic postoperative CA results in nutritional debility to the patient and as a result, patients were unable to start adjuvant chemotherapy, thus significantly affecting their disease-free survival rate.[13]

Conclusion

Injury to cisterna chyli lymphatics during abdominal surgery, which contain lymphocytes and immunoglobulin, leads to Chylous Ascites. Loss of lymphatic fluid results in lymphocytopenia, which renders patients susceptible to infection-related complications. Despite an immediate discovery of injury to cisterna chyli and repair, it had a poor prognosis and subsequent death. Further studies should consider how to recognize such an accident intraoperatively and how it may best be managed intraoperatively.

Ethical Approval

Approval to conduct this study was obtained from King Hussein Medical Centre, Amman, Jordan. Administrative clearance was obtained from the office of King Hussein Medical Centre. Written informed consent was obtained by the patient for publication of this case report and accompanying identifiable features. We also obtained informed consent for reviewing the medical files of all 30 patients who had elective pancreatic resections at KHMC and to be used as part of this medical research.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the idea, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in conscripting, revising the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be answerable for all features of the work.

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